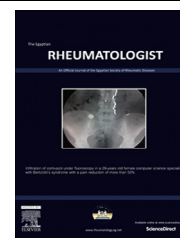




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CASE REPORT

Clinical characteristics and outcome of young West African patients with Bertolotti's syndrome



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KEYWORDS

Low back pain;
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Abstract *Aim of the work:* To assess the clinical characteristics and evolutionary track of Bertolotti's syndrome in the rheumatology department of the National Teaching Hospital Hubert Maga of Cotonou, Benin.

Patients and methods: A prospective study of 3 years (January 2011 to December 2013) evaluating the young patients suffering from Bertolotti's syndrome met in the rheumatology department in NHU of Cotonou. Patients with the syndrome were recruited and followed-up over the study period. The clinical characteristics and therapeutic outcome of the patients were assessed over a period of six months.

Results: Of 2048 patients seen during the period in the rheumatology department, 21 (1%) had Bertolotti's syndrome. The male:female ratio of the patients was 0.6 and the average age was 25.74 ± 9.32 [19–35] years. Three patients came from the neighbouring countries (one from Togo and two from Nigeria). The average disease duration was 541 ± 113 [180–656] days. The clinical symptom was dominated by low back and buttock pains (15 cases). In seven cases, there was S1 nerve root pain. Tenderness over the lumbo-sacral junction and spine was the main sign on physical examination. The evolution was satisfactory with a decline in pain and need for analgesics and NSAIDs after six months in 7 cases while a corticosteroid infiltration under fluoroscopy was required in 14 patients.

Conclusion: Bertolotti's syndrome should be considered as a separate entity in the differential diagnosis of mechanical low back pain in young patients. Good knowledge of this syndrome could contribute to improved management of low back pain.

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1. Introduction

Bertolotti's syndrome, described in 1917, refers to the occurrence of lower back or buttock pain induced by the presence of a transverse process of the mega-mobile last vertebra which

contacts the sacrum and/or in the iliac wing sometimes forming an equivalent joint space (sometimes called overly “third sacroiliac”) [1]. It affects 5–7% of the population and is found in 10% of low back pain patients aged less than 30 years [2,3]. Several cases have been reported in the Western literature review [3–11]. African literature on this syndrome is poor [12] probably related to a lack of literature on this subject.

The objective of this study is to clarify the clinical characteristics of Bertolotti's syndrome and to assess the therapeutic management in the rheumatology department of an underdeveloped country like Benin.

2. Patients and methods

This is a prospective study including patients in the rheumatology department of the National Teaching Hospital “Hubert Koutoukou Maga” of Cotonou followed-up for lumbo-sacral buttock pain ‘pygalgia’ with a mega-L5 transverse process.

The patients regularly following-up in the rheumatology department of CNHU-HKM (Cotonou) were recruited over a period of 3 years, from January 2011 to December 2013 through the following criteria: Age between 18 and 35 years; clinically presenting with lumbago, pygalgia or lower back or buttock pain isolated or associated with radiation along the S1 nerve root; having an anteroposterior plain radiograph of the lumbar spine showing a mega-apophysis of L5 with sacroiliac neo joint-related clinical symptoms and received the prescribed therapeutic.

Patients with painful symptoms associated with another obvious aetiology were not included in the study. The study was approved by the local ethics committee and conforms to the 1995 Helsinki declaration. All patients gave their informed consent prior to their inclusion.

The demographic parameters (age, gender), disease duration, the clinical symptoms, treatment and scalable data were collected with a structured sheet survey.

Analgesics used were those of levels I (paracetamol) and II (paracetamol or tramadol in combination with codeine or caffeine) for a period of 15 days and maintained on demand throughout the duration of the study. Non-Steroidal Anti-Inflammatory Drugs (NSAIDs) (diclofenac, ketoprofen and celecoxib) were used at a maximum dose for 15 days. No NSAIDs were given later. In cases with severe pain, fluoroscopy guided injections were considered.

The visual analogue scale (VAS) of pain of the patients was evaluated at the 3rd and 6th months. The primary criterion was the reduction in the VAS.

Statistical analysis: Data were analysed using the Office excel software. Results were presented as mean \pm SD and number and percentage.

3. Results

3.1. Socio-demographic characteristics

Out of 2048 patients seen during the period of the study, 21 (1%) had Bertolotti's syndrome. The syndrome represented 2.8% of LBP outpatients. The sex (male: female) ratio was 0.6. The average age of patients was 25.74 ± 9.32 [19–35] years. Three patients came from the neighbouring countries (one from Togo and two from Nigeria).

3.2. Clinical features

The patients' clinical presentation was essentially unilateral (20 cases) with average disease duration of 541 ± 113 [180–656] days. The clinical symptom was represented by low back pain and lumbar-buttock pain. There was an ipsilateral radiation along S1 with musculoskeletal injury in 7 patients (Table 1). Tenderness over the para-spinal muscles next to the lumbo-sacral junction and painful extension of the spine were the main physical examination signs present in all patients. The plain radiograph of the lumbar spine showed a mega-apophysis of L5 with the sacroiliac joint. (Figs. 1 and 2).

3.3. Therapeutic data

Apart from the analgesics and NSAIDs used by almost all patients, 67% have used a series of 3 fluoroscopy guided injections (one week apart) of cortivazol for pain relief (Fig. 3). Table 2 summarizes the therapeutic data (see Table 3).

3.4. Outcome

The evolution was satisfactory in 14 patients with a decline of pain after six months (residual pain less than 50% and a total absence of drug consumption). Three patients had continuous pain (despite a series of 3 fluoroscopy guided injections). Four patients had a relapse after 6 months which required a repeat of infiltration series.

4. Discussion

Bertolotti's syndrome is a mega-transverse process of the last lumbar vertebra provoking a neo-articulation with the sacrum and the ilium. It is a syndrome that is not an uncommon cause of back pain in young adults and may be a source of chronic pain in 10% of LBP patients younger than 30 years [4,5,12]. The hospital frequency of this pathology and the young age of the patients observed in our study attest to the cosmopolitan character of this syndrome in young adults.

On the pathophysiological level, two mechanisms have been proposed to explain the pain. The work teams Pekindil et al. [13] and Connolly and coworkers [14] suggest a direct mechanism related to the suffering of neo-articulation detected by SPECT-CT in symptomatic patients. On the other hand, other studies such as those of Luoma et al. [15], Bron et al. [16] and Aihara and associates [17] suggest a rather indirect mechanism by premature wear of the L4–L5 disc due to improper locking of the iliolumbar ligaments.

Table 1 Clinical symptoms in the patients with Bertolotti's syndrome.

	N	(%)
Lateralized LBP	8	(38)
Low back/buttock pain	5	(23)
Ipsilateral buttock pain	1	(6)
Sciatica	7	(33)
Neurologic deficit	0	(0)



Figure 1 Right mega-apophysis of L5 forming a new transverse iliac joint in a 24 year old student.

Clinically, all of our patients had LBP or lumbar-buttock pain. The symptoms may be clinically presented as a sacroiliac disease [18]. In the present study, pseudo-root involvement was present in one third of cases in contrast to the series of Avimadje et al. [2], where two-thirds of their patients had sciatica. Shibayama et al. suggested that sciatica syndrome may result from extra-foraminal nerve compression caused by the enlarged transverse process [19].

Infiltration under fluoroscopy of cortivazol was given to the majority of patients in our series with a pain reduction of more



Figure 2 Bilateral Bertolotti's syndrome in a 28 year old banker.



Figure 3 Infiltration under fluoroscopy of the Bertolotti's syndrome in a 29-year old female computer science specialist.

Table 2 Therapeutic data in the patients with Bertolotti's syndrome.

	N	(%)
Analgesics	21	(100)
Non Steroidal Anti-Inflammatory Drugs	20	(95)
Short term oral corticosteroids	2	(9.5)
Fluoroscopy guided corticosteroid injections	14	(67)
Surgery	0	(0)

Table 3 Outcome in patients with Bertolotti's syndrome.

	Residual pain < 50% N (%)	≥ 50% N (%)
After 3 months	18 (85.7)	3 (14.3)
After 6 months	14 (66.7)	7 (33.3)

than 50% in more than half of all patients after six months. This is in agreement with the study of Avimadje et al. [2] in

France where infiltration was the main treatment used. A literature review revealed that surgery is often used for the treatment of this condition especially in refractory cases to medical treatment. It is often excisions of the mega- apophysis or arthrodesis with questionable efficacy [20–22].

Proposed in the 3 cases in our series with continuous non declining pain after one month, surgery could not be done due to lack of financial resources and fear to undergo spine surgery. Moreover, Delisser and Burducea proposed a radio-frequency treatment to enhance the beneficial effect of infiltration [23].

In conclusion, Bertolotti's syndrome should be considered as a separate entity in the differential diagnosis of mechanical low back pain in young patients. Its diagnosis is based on the simple presence of a transverse mega-process of the last lumbar vertebra, which articulates with the sacrum and/or iliac wing forming the source of pain. However, the diagnosis is done by exclusion after ruling out all other causes of back pain. The number of cases reported in our series shows the importance of this syndrome in Benin and West African countries. Infiltration of corticosteroids under fluoroscopy remains an effective treatment for the management of this syndrome.

Conflict of interest

The authors declare having no conflict of interest related to this research work.

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